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CASE REPORT

Transformation of a meningioma with atypical imaging

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ABSTRACT

Meningiomas are benign tumors of the central nervous system. They have long term curability if they are excised completely. If not, they can recur after a prolonged period and can lead to increased morbidity during re-surgery. Recurrence is rarely associated with invasiveness. Usually de-differentiation in case of meningiomas is uncommon without any predisposing factors including different genetic mutations or radiation to the involved region. We report a case of a 38-year-old female who was operated for a benign para-sagittal meningioma 8 years back and subsequently developed an invasive recurrence off late. Also this time, the imaging morphology was slightly different for a meningioma and gross as well as microscopic findings were very atypical. Awareness for such cases must be there while dealing with recurrent meningiomas as invasiveness may not always be associated with adverse predisposing factors like radiation. As invasiveness is always a histopathological diagnosis, picking up such features on imaging is a daunting task and if done, can help neurosurgeons prognosticate such invasive recurrences in a better fashion.

Key words: Invasive, meningioma, recurrence, transformation

Introduction

Meningiomas have been intriguing since the times of Sir Harvey Cushing and still they can be easily the most difficult tumours to excise completely. They have characteristic radiological findings as usually they are iso-intense on T1-weighted images, iso-hypointense on T2-weighted images and are intensely enhancing post gadolinium. Recurrence is rarely associated with invasiveness without any predisposing factors like prior radiation exposure. We had a 38-year-old female who was operated for a benign para-sagittal meningioma 8 years back. She had a recurrence after 8 years and this time, the tumour was having an entirely different morphology, both grossly as well as microscopically.

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Case Report

A 38-year-old lady presented to us with severe headache and vomiting 8 years back. She had no neurological deficits. On imaging a para-sagittal meningioma was diagnosed. She underwent left frontal craniotomy and complete excision of the meningioma was performed [Figure 1]. The histopathology was grade I transitional meningioma. She was under regular follow up and several interval scans did not reveal any recurrence [Figure 2]. Recently, after a span of 8 years, she had a seizure episode when magnetic resonance imaging (MRI) showed a recurrent lesion in the same anatomical location which was iso-intense on T1-weighted images, extremely hyperintense on T2-weighted images and showed scanty contrast enhancement with lobulated margins [Figure 3]. These imaging findings were quite atypical for a meningioma. Subsequently, she underwent a second surgery where, the tumor had a mucoid "jelly" like consistency with ill-defined margins and there was no distinct arachnoid plane between the tumor and normal brain. The tumour was sucked out like a viscous fluid with relative ease, although it was difficult to demarcate the peripheral margins. The overlying bone was also infiltrated by the tumour [Figure 3]. This time the histology progressed to grade II atypical invasive meningioma [Figure 4]. The Ki 67 index was moderate and vimentin and epithelial membrane antigen were found to be positive. Eighty percent of cells had positive results for estrogen and progesterone receptors. The patient had an uneventful post operative stay and she was later on subjected to radiotherapy and is doing well in follow-up.

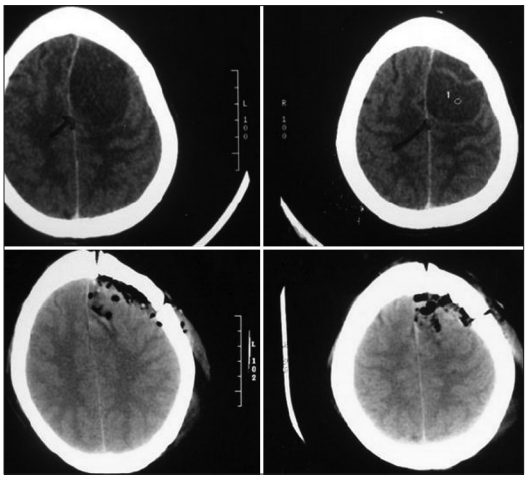

Figure 1: Initial computed tomography scan demonstrating pre and post operative left para-sagittal meningioma 8 years back

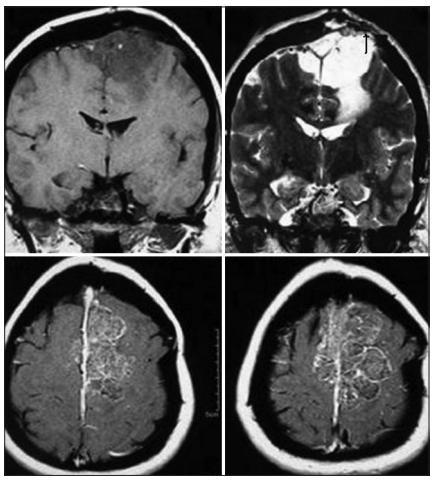

Figure 3: Coronal and axial views of magnetic resonance imaging brain showing a parasagittal lesion which is isointense on T1 weighted images; extremely hyperintense on T2 weighted images and showing scanty contrast enhancement with lobulated margins. Bone invasion can be also be appreciated (arrow)

Discussion

This case of transformation in a benign meningioma into an invasive variety without any triggering factors was thought provoking. The rate of malignant transformation in meningiomas is around 2%.[1,2] Losses of 1p, 9q, 10q and 22q have been correlated with such type of de-differentiation. Out of these, loss of 22q has been associated with higher percentage of grade II and III meningiomas. We could not find any report of an invasive meningioma with such atypical imaging characteristics in the literature as invasiveness is essentially a histological diagnosis. Brain invasion has been defined as the loss of leptomeningeal interface between meningioma and Glial Fibrillary Acidic Protein stained brain parenchyma. There has been a mention of occasional loss of cerbro-spinal fluid rim around the tumor on MRI in five cases by Nakasu *et al.*[3] Out of these five, two turned out to be invasive meningiomas. The recurrence rates of grade II

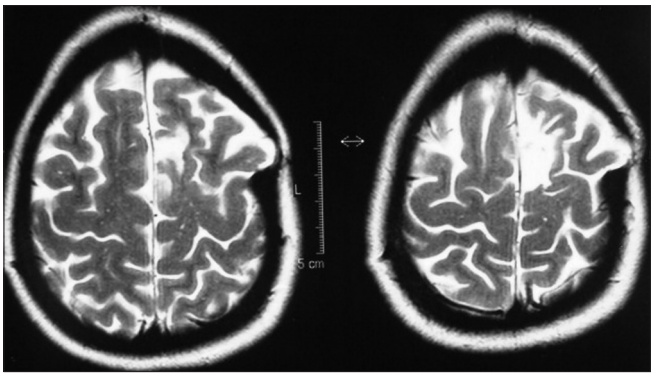

Figure 2: Interval scan showing no recurrence 4 years back

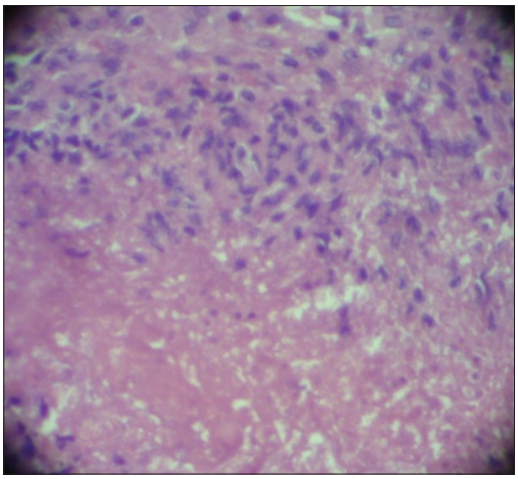

Figure 4: Histopathology showing meningothelial cells with evidence of fibrinoid necrosis suggestive of high grade tumor (×100)

(atypical) meningiomas range between 29% and 52%.[4] Almefty *et al.* had detected malignant progression in 11 (6.2%) patients in their landmark series of 175 recurrent meningiomas.[5] All the progressed tumours had an aggressive clinical course. They concluded that increased MIB index and deletions of 22, 1p, and 14q were common in the progressed tumors. LeMay *et al.* reported a case where a benign meningioma transformed into malignancy after being operated upon for 11 times for subtotal resections over a period of 18 years.[6] This patient later on went on to develop a pulmonary metastasis. Ohba *et al.* reported a malignant transformation in a 57-year-old woman with meningothelial petro-clival grade I meningioma within a span of 1 year.[7] The MIB-I index and p53 positivity were 4.6% and 35.4% respectively during the primary surgery which showed an increase upto 34.7% and 33.1% during the second. Our case also had moderate to high proliferation index, although these chromosomal deletions were not found. Also, loss of progesterone receptors has been associated with an aggressive course, although this was not seen in our patient. We could not establish the role of SPARC (secreted protein acidic and rich in cysteine) protein in our case. This protein has been studied extensively as a marker of invasive meningiomas.[8] It's an extracellular matrix associated protein implicated in the modulation of cell adhesion

and migration. It is helpful in differentiating histologically benign invasive tumors from histologically benign non-invasive tumors and is found invariably in all invasive meningiomas.

Hence, there were two strange findings in this case. One, the gross and radiological appearance was quite atypical for a meningioma and secondly, the transformations of a benign pathology to an invasive one over a period of 8 years without any predispositions like prior radiation. Invasive meningiomas may present with atypical imaging characteristics and one must keep in mind this variation in the image morphology, as it may point towards a peculiar and possibly a non favorable histology. Hence, it could lead us to predict unfavorable histology on imaging. Although further insight can be gained by analyzing few more cases of such invasive meningiomas and then only definitive conclusions may be derived regarding its real clinical implications.

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